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Case report

Idiopathic bronchocentric granulomatosis in an asthmatic adolescent

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ABSTRACT

Bronchocentric granulomatosis in asthmatic patients has been generally considered to be associated with allergic bronchopulmonary aspergillosis and represent a histopathologic manifestation of fungal hypersensitivity. Here we report a case of an idiopathic bronchocentric granulomatosis in a 17-year-old man with a history of asthma. He was admitted to the hospital with a fever and cough, and a chest CT scan showed peribronchial consolidation in the pulmonary parenchyma, which was unresponsive to antibiotic therapy. The pathological findings obtained by video-assisted thoracoscopic lung biopsy revealed necrotizing granulomatous inflammation centered on bronchi and bronchioles and there was no evidence of fungal colonization, resulting in a diagnosis of idiopathic bronchocentric granulomatosis. Systemic corticosteroid therapy led to clinical and radiological recovery. Physicians should take into account the possibility of the idiopathic process in bronchocentric granulomatosis of asthmatic patients.

1. Introduction

Bronchocentric granulomatosis (BCG) is a histological diagnosis characterized by a granuloma formation and necrosis affecting bronchi and bronchioles [1,2]. BCG has been described in association with asthma and allergic bronchopulmonary aspergillosis (ABPA), whereas the cases of non-asthmatic patients with BCG are usually idiopathic [2,3]. Here, we report a case of idiopathic BCG in an asthmatic adolescent without accompanying pathological aspergillus infection.

2. Case report

A 17 year-old man presented with fever and non-productive cough lasting for more than 1 week. Although he had a history of asthma in childhood, his asthmatic symptoms had remitted and he used a short-acting bronchodilator only occasionally in the last few years. Upon examination, his oxygen saturation was 98% on room air and no crackles nor wheezes were auscultated over the chest. Laboratory investigation showed an increased white blood cell count of 13890/mm^3 with 82.9% neutrophils and 0.4% eosinophils. The level of C-reactive protein had also increased to 5.00 mg/dl. Serum IgE level was slightly elevated to 633 IU/ml. A high resolution CT scan demonstrated focal consolidation in the left lower lobe without the evidence of central bronchiectasis or mucoid impaction (Fig. 1A). We diagnosed as community acquired bronchopneumonia and oral azithromycin was administrated as antibiotic therapy. However, he was admitted to our hospital 6 days later, suffering from prolonged fever. Rapid influenza antigen test was negative for influenza A and B. The galactomannan antigen of Aspergillus, antibodies of mycoplasma and human-immunodeficiency virus were negative. Serological tests for antinuclear cytoplasmic antibody and rheumatoid factor were also negative. The levels of angiotensin-converting enzyme, (1–3)-β-D glucan were normal. The level of soluble interleukin-2 receptor in serum was slightly elevated to 613 U/ml.

He was started on an intravenous infusion of sulbactam/ampicillin for the first three days followed by administration of meropenem plus clindamycin. However, his fever persisted and the re-evaluation of the CT scan on day 4 after hospitalization revealed the exacerbation of left lower lobe consolidation, and new infiltrative opacity appeared in the right upper lobe (Fig. 1B). Bronchoscopy results including mycobacterial and fungal culture were negative and revealed no malignancy. He then underwent video-assisted thoracoscopic lung biopsy of the right upper lobe on day 11, because we considered the possibility of lymphomatoid granulomatosis, malignant lymphoma and granulomatosis with

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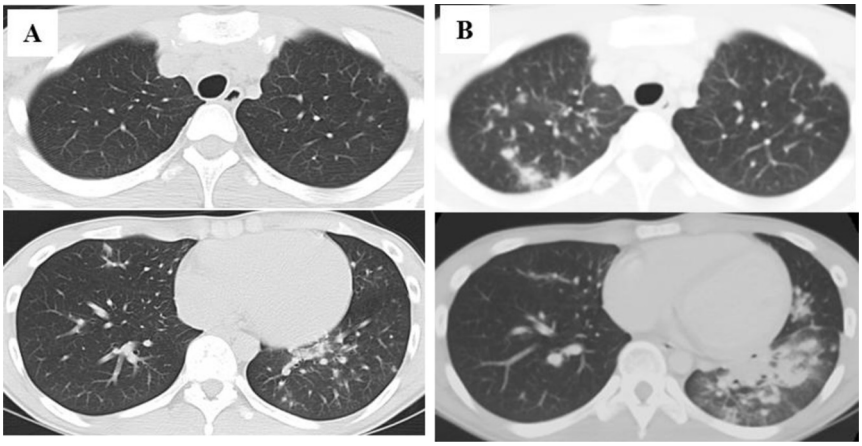

Fig. 1. (A) A chest CT scan on admission. Focal consolidation in the left lower lobe is confirmed. (B) A chest CT scan on day 4 after hospitalization. Appearance of consolidation in the right upper lobe and the deterioration of the lung shadow in left lower lobe can be seen.

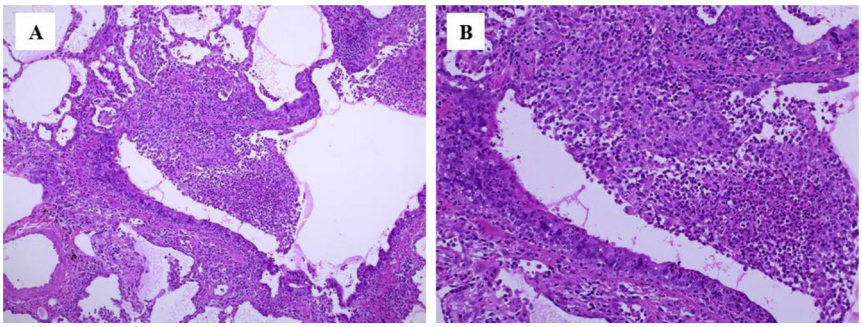

Fig. 2. Histopathologic examination (Hematoxylin and eosin stains) of lung biopsy specimen. (A) The bronchiolar mucosa is partially replaced by necrotizing granulomatous inflammation. The cellular infiltrate is present in the lumen and the peribronchiolar lesion. (B) Higher magnification photomicrograph shows granulomatous inflammation and remnant of respiratory epithelium.

polyangiitis based on his clinical course and imaging findings. The pathology of the resected specimen revealed necrotizing granulomatous inflammation centered on bronchi and bronchioles (Fig. 2). Histiocytes, neutrophils and lymphocytes infiltrated in the affected airways, but eosinophils were hardly observed. The Grocott and periodic acid-Schiff (PAS) stains showed no fungal elements and Ziehl-Neelsen stain was also negative. The pathology was diagnostic of BCG, resulting in a diagnosis of idiopathic BCG based on no obvious evidence of the underlying disease. He was prescribed oral prednisolone 30 mg daily with rapid resolution of the fever. The improvement of radiograph findings also confirmed and oral steroids were gradually tapered. He had remained free of relapse for six months after corticosteroid withdrawal.

3. Discussion

Katzenstein et al. described two patterns of BCG based on the presence or absence of asthma [3]. Approximately half of all cases are related to asthma and the prevalence appears to be increased in younger age. Patients in this group generally manifest with pulmonary complaints such as cough, dyspnea and wheezing. BCG in asthmatic patients has been regarded as a form of ABPA and a hyperimmune response to colonizing fungi in the airway [3–5]. On the other hand, non-asthmatic patients with BCG tend to be older and less symptomatic. This group lacks evidence of fungal hypersensitivity and is considered to be generally idiopathic, although it has been reported to be associated with numerous diseases such as rheumatoid arthritis, granulomatosis with polyangiitis, pulmonary echinococcosis, and mycobacterial/fungal infection [5].

Our patient had asthma in adolescence and developed chest symptoms in onset, which typically presents in the asthmatic group with BCG. However, we considered that this case differs from the condition of ABPA, because fungal hyphae and tissue eosinophilia were not identified in the involved airways of lung biopsy specimen [3,6]. Furthermore, we were unable to determine the etiology of BCG other than fungal infection. This case of BCG is idiopathic in an asthmatic patient and extremely rare condition. We speculate that there would be excess immunoreactions with unidentified exogenous agents, because the clinical course was rapidly progressive and there was no clear evidence of the underlying disease.

Corticosteroids are generally effective for BCG, although some cases require long-term therapy due to recurrent or persistent disease [3,4]. Our case was successfully treated with corticosteroid monotherapy. It is very important to search the underlying condition that is associated with BCG before starting to administer corticosteroids. In particular, ABPA is recommended to administer antifungal agent with corticosteroids [7–9]. In this case, lung biopsy by video-assisted thoracoscopy helped to exclude the possibility of fungal infection, and avoid unnecessary administration of antifungal agents. We consider that BCG in asthmatic patients may occur in not only fungal hypersensitivity but also idiopathic process without fungal infection.

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